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Original Article

The impact of the COVID-19 pandemic on the emotional well-being and home treatment of Belgian patients with cystic fibrosis, including transplanted patients and paediatric patients



Trudy Havermans^{a,*}, Janne Houben^a, Francois Vermeulen^b, Mieke Boon^b,
Marijke Proesmans^b, Natalie Lorent^c, Erik de Soir^d, Robin Vos^c, Lieven Dupont^c

^a Clinical Psychologist, University Hospitals Leuven, Leuven, Belgium

^b Pediatric Pulmonologist, University Hospitals Leuven, Leuven, Belgium

^c Pulmonologist, University Hospitals Leuven, Leuven, Belgium

^d Clinical Psychologist, Royal Higher Institute for Defence, Department of Scientific and Technological Research, Brussels, Belgium

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ABSTRACT

Background: Little is known about the impact of COVID-19 on patients with cystic fibrosis (CF), despite being considered a high-risk group. This study explored the early impact of COVID-19 on the emotional well-being of patients and self-reported changes in their home therapy since the start of the pandemic.

Methods: Adult patients with CF, lung-transplanted (LTX) CF patients and parents of children with CF completed an online questionnaire, securely linked to their medical files. The questionnaire covered the emotional impact of the pandemic, changes in CF and LTX treatment, changes in health-protecting behaviours and CF-related concerns, and their perception of their COVID-19 status.

Results: The response rate was 63% (80 CF, 66 LTX and 73 parents). A wide range of illness severity was included. None of the respondents had contracted COVID-19 and all strictly followed the social distancing rules. There was evident psychological impact, with many reporting increased stress, fear and worry about CF and the future. Changes in treatment were positive, including more physiotherapy for adults and better-quality nebulizing. Changes in routine were reported, such as different treatment timing. Adult patients and parents had cancelled their CF appointments more often since the start of the pandemic.

Conclusions: The initial psychological impact of COVID-19 was evident. The impact on home treatment was reassuringly small. Psychological care is needed for patients suffering prolonged psychological impact, and CF teams need to contextualize the information that patients and parents receive from the media and support them to balance the perceived risk with true risk.

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1. Introduction

Cystic Fibrosis (CF) is the most common life shortening genetic disorder with a worldwide prevalence of about 1 in 4000 births. CF is a multi-organ disease, affecting lungs and digestive system. The daily intensive treatment has multiple components and is considered a heavy burden on patients and their families. Although

life expectancy has increased over the past decade and is expected to improve further with promising CFTR modulators, no cure is presently available. Thus far, lung transplantation (LTX) is the only option to overcome respiratory insufficiency, but LTX involves further chronic comorbidities requiring intensive treatment. Studies have shown that multidisciplinary and centre care are effective in improving both longevity and the quality of life of people with CF [1]. Since the start of 2020, the SARS-CoV-2 pandemic (COVID-19), has had a worldwide impact on health care facilities, the economy and social life.

1.1. Self-isolation during COVID-19

As yet, little is known about the impact of COVID-19 on chronically ill patients, even though they are considered a high-risk

Abbreviations: BMI, body mass index; CF, cystic fibrosis; CFTR, cystic fibrosis transmembrane conductance regulator; CLAD, chronic lung allograft dysfunction; COVID-19, coronavirus disease 2019; FEV₁ %pred, percent predicted forced expiratory volume in 1 second; LTX, lung transplantation; SARS-CoV-2, severe acute respiratory syndrome coronavirus 2.

* Corresponding author.

E-mail address: trudy.havermans@uzleuven.be (T. Havermans).

group; the available information has mainly been extrapolated from data about increased risk in the context of influenza [2]. Patients with CF, and especially LTX patients, because of their suppressed immune system, are advised to actively prevent getting COVID-19, through hygiene precautions, social distancing and self-isolation.

A recent review of the literature on the psychological impact of quarantine on healthy people retrieved reports on negative psychological effect, including post-traumatic stress symptoms, confusion, and anger. Stressors include longer quarantine duration, fear of infection, frustration, boredom, inadequate supplies (e.g. food), inadequate information, financial loss and stigma [3]. Several risk factors associated with the development of post-traumatic psychological illnesses have been identified [4], including the duration of the event, previous psychological problems and acute stress disorder. Predictors of later psychological problems include: feeling one's life is threatened, feelings of shame or blame towards others, appraisal of the traumatic event as uncontrollable or unpredictable, a history of previous trauma, a lack of accessible social support, and alcohol abuse or increased smoking. Patients with CF have an increased risk for mental health problems due to their chronic illness [5], putting them at risk for developing further psychological problems during the pandemic.

A national lockdown was enforced by the Belgian government on March 18, 2020. People had to stay indoors as much as possible which meant they had to work from home. Schools and public places were closed, only food stores and pharmacies were opened to a limited number of people at the same time and non-essential movements, except for walking, running and cycling, were forbidden.

The COVID-19 outbreak has disturbed regular patient follow-up. The CF and Lung transplant centre at our hospital were required to comply with hospital restrictions implemented in preparation for the expected admissions of patients with severe COVID-19 infection and in accordance with the COVID-19 contingency measures. These restrictions included the cancellation of routine outpatient clinics, as well as most planned hospitalizations, investigations and treatments, which are very much part of CF and LTX care. We organized our multidisciplinary care differently, for example through telephone or video consultations. The impact of these changes on the multidisciplinary care of patients with CF is unclear, although we expect that the cancellation of routine CF clinics and monitoring, as recommended by the ECFS standards of care [6], will have a negative impact on CF prognosis and patients' well-being over time.

The aim of this study was to explore the short-term impact (within 4–6 weeks of lockdown) of the COVID-19 pandemic on the emotional well-being of patients with CF, including those who underwent LTX, and to record self-reported changes in home treatment due to the pandemic. Previous studies have shown an association between psychological symptoms or quality of life in both patients and parents and severity of illness [5,7]. We therefore hypothesized that an increase in self-reported emotional problems in the context of the COVID-19 pandemic is associated with lower lung function (measured as FEV₁%pred), lower body mass index (BMI) or the presence of Chronic Lung Allograft Dysfunction (CLAD) (for LTX patients). In addition, we hypothesized that lower FEV₁%pred, BMI or CLAD status is associated with greater changes in treatment (indicative of adherence to treatment).

Responses to a self-administered questionnaire, distributed via eHealth, were analysed to test these hypotheses.

2. Methods

2.1. Patients and parents

All patients ($n = 375$; 151 adults, 132 LTX, 92 children) undergoing regular follow-up at the CF centre and the Lung transplant centre of the University Hospitals Leuven, Belgium were considered for participation. To establish exclusion criteria, the CF team considered the potential burden of the study on our patient population. The following exclusion criteria were applied for patients (16 years and older) and parents (of children in the ages 1–18 years): non-Flemish speaking, a history of mental illness (psychiatric treatment for severe clinical anxiety and/or depression), a history of learning difficulties, less than 1 year since CF diagnosis, less than 1 year since LTX, current hospitalization, and age above 70 years of age. In families with more than one child with CF, the parents were asked to participate once, referring to their oldest child with CF.

As a result, 16 of 151 adults with CF were excluded (language ($n = 6$), learning difficulties ($n = 2$), history of mental illness ($n = 2$), current hospitalization ($n = 1$), recent diagnosis ($n = 1$), or age above 70 years [4]). Twenty-five of 132 LTX patients were excluded (language ($n = 5$), LTX < 1 year ($n = 10$), age above 70 ($n = 2$) or current hospitalization ($n = 8$)). Twelve parents of 92 children were excluded (recent diagnosis ($n = 3$), history of mental illness ($n = 3$), learning difficulties in the parents ($n = 2$) or sibling with CF ($n = 4$). Consequently, an invitation for the study and a link to the questionnaire was sent to 322 of 375 patients or their parents. The demographic data collected for each patient included age, sex, family composition during the lockdown, work/study situation during this period and medical variables extracted from the patient files, including lung function, BMI, or BMI z-score for the children, and CLAD status for LTX patients.

2.2. Procedure

The study period started three weeks into the lockdown, lasting from April 10 to April 23, 2020. During this period, 13,786 confirmed COVID-19 diagnoses, 3862 hospitalizations and 3103 deaths from COVID-19 were reported in Belgium (<https://epistat.wiv-isp.be/covid/covid-19.html>). The patients and parents were informed about the study by mail. They were then sent a link to the questionnaire via the MyNexuzhealth application. This secure application is linked to the hospital's electronic medical patient records, allowing patients and parents to access their medical information. By completing the questionnaire, the patients/parents implicitly agreed that their pseudonomized data would be used for study purposes. This study was approved by the Ethics Committee of the University Hospitals Leuven (S63974).

2.3. Questionnaire

Two versions of the questionnaire were constructed, one for patients and one for parents. Both were in the Flemish language. The content of both versions was identical. Parents reported about themselves and on changes in their child's treatment. For the present article, the items have been translated into English by two of the authors (TH and JH), but the translations have not been validated. With no previous references available, the CF and LTX teams decided that a cut-off frequency > 30% was considered noteworthy.

Table 1
Demographic and medical variables of the patients with CF and parents.

	Adult with CF (n = 80)	CF with LTX (n = 66)	Parents of children with CF (n = 73; of 59 children)
Age, years; mean (SD) range	32.2 (10.7) 16–65	39.4 (10.1) 21–67	39.6 (5.7) 26–53
Male - Female (Mother-Father) n	42–38	25–41	19–54
Age child, years; mean (SD) range	NA	NA	9.2 (4.2) 1–16
Boys/girls n	NA	NA	25–34
Number of adults in the house during lockdown; range	1–5	1–4	1–4
Number of children in the house during lockdown; range	0–2	0–3	1–5
Work/school situation during lockdown			
Disability due to CF	15%	51.5%	NA
Working from home	17.5%	1.5%	48.5%
Home schooling	15%	7.6%	NA
Working outside the home	2.5%	3.0%	16.2%
Sick leave due to COVID-19	8.8%	18.2%	4.4%
Unemployed due to COVID-19	12.5%	4.5%	11.8%
Other (including missing data)	28.8%	13.6%	19.1%
Medical variables			
FEV ₁ % pred; mean (SD) range	70.4 (22.53) 21–110	79.5 (20.11) 22–126	98.0 (16.2) 65–142*
BMI, kg/m ² , or BMI z-score (for children); mean (SD) range	22.5 (30.1) 15.7–31.4	20.5 (32.1) 13.2–29.3	–0.19 (0.7) –1.6–1.9
Chronic Lung Allograft Dysfunction status; yes	NA	15	NA

FEV₁, Forced expiratory volume in 1 s; BMI, Body Mass Index.

* 15 children not able to perform spirometry. CF, Cystic Fibrosis; LTX, lung-transplanted patients.

2.3.1. Emotional impact

The first part of the questionnaire included items relating to emotional well-being of the adult patients and parents during the lockdown. No appropriate pre-existing questionnaire was available and so we based the items on questionnaires for health care providers working in traumatic circumstances, which have recently been used for health care workers involved in the care of patients with COVID-19 [8]. The author gave us written permission to adapt the questionnaires for use in the CF population.

All the questions related to COVID-19 and were rated on a 3-point scale: 'not at all', 'sometimes' or 'often'. The questions covered 36 stress related descriptions; these described individual items or subsets of items referring to concepts such as overall tiredness or loss of routine. One item asked about suicidal thoughts. When a respondent replied 'sometimes' or 'often' to this item, he or she was contacted by phone as soon as possible, and support or referral were offered. In line with the procedure suggested by de Soir, the scores were recoded into two groups, 'not more' since the start of the COVID-19 pandemic and 'more' since the start of the COVID-19 pandemic.

2.3.2. Home CF and LTX treatment

The second part of the questionnaire was designed to identify self-reported changes in home CF and LTX treatment, since the start of the COVID-19 pandemic. Changes in four treatment modalities were examined: oral medication, nebulized therapy, physiotherapy and nutrition. Changes in adherence to treatment can be related to stress, lack of routine or attitudes and beliefs about the treatment [9]. The patients and parents were asked about unintentional changes (such as forgetting treatment) and intentional changes (such as doing more, skipping a treatment, doing better, different timing, or doing less) in their home treatments during the COVID-19 pandemic. Again, the respondents were asked to rate the items on a three-point rating scale ('never', 'sometimes' or 'often') and the scores were recoded into two groups, 'not more' and 'more' since the start of the COVID-19 pandemic. Finally, three questions were asked about visits to or by the home physiotherapist, who were advised by the Belgian government to limit their (home) visits.

2.3.3. Changes in health-protecting behaviours and CF related worries

In the third part of the questionnaire, the patients and parents were asked about changes in their behaviours or worries about CF

since the start of the COVID-19 pandemic. This part included 4 items about social distancing (e.g. 'I/my child stay(s) at home all the time'), 6 items about health-protecting behaviours (e.g. 'I/my child take(s) more vitamins'), 1 item about exercise ('I/my child exercise(s) more'), and three items about CF related worries (e.g. 'I worry more about my CF/my child's CF'). As before, the respondents rated the items on a 3-point scale 'never', 'sometimes' or 'often'. However, for these items the scores were recoded into the two groups, 'no' and 'yes' since the start of the COVID-19 pandemic.

2.3.4. COVID-19 status

In the final part of the questionnaire, the patients and parents were asked to tick one of four boxes describing their perception of whether they or their child currently or previously had COVID-19, and whether they had been tested.

2.4. Statistical analyses

SPSS version 26 was used for statistical analyses. Frequencies were calculated and Chi-square and ANOVA analyses were used to explore selected univariate associations between subgroups.

3. Results

3.1. Demographic and medical data

A total of 80 adults (58%), 66 LTX patients (61%) and 73 parents of 59 children (73%) completed the questionnaire (supplementary Table 1). Table 1 shows demographic and medical characteristics. Thirteen percent of the CF patients reported they were alone during the lockdown and 63% did not live with children, whereas 29% of the LTX patients were alone and 81% were not spending the lockdown with children. Around 65% of the parents reported that they were spending the lockdown with one other adult. The medical data showed that the sample included a wide range in illness severity.

3.2. Emotional impact

Table 2 summarizes the responses to the items relating to the emotional impact experienced since the start of the COVID-19 pandemic. Three respondents reported an increase in suicidal

Table 2

Responses on the emotional impact on patients with CF since the start of the COVID-19 pandemic, classified as not more or more than before the pandemic.

	Patients with CF n = 79 (%)		CF with LTX n = 64 (%)		Parents of children with CF n = 72 (%)	
	Not more	More	Not more	More	Not more	More
Overall tiredness						
I have no energy at the end of the day	61.5	38.5	62.9	37.1	56.2	43.8
I already have no energy at the start of the day	80.8	19.2	72.6	27.4	80.8	19.2
Parts of my body feel weak	71.8	28.2	74.2	25.8	75.3	24.7
I have little energy reserve and I am near breakdown	85.9	14.1	85.5	14.5	83.6	16.4
My arms and legs feel heavy	87.2	12.8	79	21	89.0	11.0
My body feels stiff and rigid	83.3	16.7	72.6	27.4	79.5	20.5
I feel like isolating myself all the time	69.6	30.4	72.6	27.4	75.3	24.7
I feel no hunger or thirst	92.3	7.7	96.8	3.2	94.5	5.5
Even the smallest tasks are demanding for me	83.3	16.7	83.9	16.1	79.5	20.5
Feelings of depression						
I feel sadness and depressed	66.7	33.3	67.7	32.3	69.9	30.1
I feel discouraged and demoralized about the future	69.6	30.4	61.3	28.7	61.6	38.4
I cannot experience pleasure	70.9	29.1	71	29	83.6	16.4
I feel helpless because I cannot help my family or friends	57.7	42.3	54.8	45.2	56.2	43.8
I have lost confidence	83.3	16.7	75.8	24.2	94.5	5.5
I am critical about myself	73.1	26.9	75.8	24.2	69.9	30.1
I am critical towards others	62.8	37.2	56.5	43.5	46.6	53.4
I have lost interest in things	94.9	5.1	87.1	12.9	89.0	11.0
I have difficulty taking decisions	84.6	15.4	79.2	21.8	79.5	20.5
I do not have much social support	88.5	11.5	85.5	14.5	86.3	13.7
I am ashamed of some of my reactions	85.9	14.1	87.1	12.9	84.9	15.1
I experience this situation as worsening (deaths, hospitalizations)	48.7	51.3	33.9	66.1	27.4	72.6
I cannot adhere to my usual routine	41	59	45.2	58.8	43.8	56.2
I suffer from previous trauma	87.2	12.8	79	21	87.7	12.3
Alcohol and smoking						
I drink more alcohol to cope	94.9	5.1	98.4	1.6	89.0	11.0
I smoke more to cope	98.7	1.3	95.3	4.7	98.6	1.4
Symptoms of post-traumatic stress						
I experience increasing levels of stress	53.8	46.2	53.2	46.8	37.0	63.0
I have negative thoughts about the crisis	60.3	39.7	61.3	38.7	61.6	38.4
I have nightmares about the crisis	83.3	16.7	75.8	24.2	83.6	16.4
I have disturbing physical reactions (heart palpitations, sweating etc.) when I think about the crisis	84.6	15.4	80.6	19.4	87.7	12.3
I have difficulty sleeping	51.3	48.7	59.7	40.3	68.5	31.5
I have difficulty concentrating	75.6	24.4	69.4	30.6	69.9	30.1
I am extra alert for situations possibly dangerous to myself or others	21.8	78.2	11.3	88.7	12.3	87.7
I am irritated with the unexpected circumstances related to the COVID-19 crisis	62.8	37.2	56.5	43.5	53.4	46.6
I am afraid of being infected by the virus	21.8	78.2	17.7	82.3	15.1	84.9
I am afraid of spreading the virus	60.3	39.7	53.2	46.8	30.1	69.9

CF, Cystic Fibrosis; LTX, lung-transplanted patients.

thoughts; all three were contacted promptly. Over 30% of the respondents reported a loss of energy at the end of the day, feeling sad and discouraged about the future, or having negative thoughts about the crisis. Over 40% felt helpless because they could not meet with family members or friends, felt more critical towards others, or experienced increasing levels of stress. Around 40% of

the adult patients (CF and LTX) and 30% of the parents reported more difficulty sleeping. More than 50% experienced the situation as worsening or reported they could not adhere to their usual routine. In addition, 80% of the respondents reported they were extra alert for possible dangerous situations to themselves or others.

Table 3

Changes in home oral medication and nutrition since the start of the COVID-19 pandemic, classified as not more or more than before the pandemic.

	Patients with CF <i>n</i> = 78 (%)		CF with LTX <i>n</i> = 63 (%)		Parents of children with CF <i>n</i> = 72 (%)	
	Not more	More	Not more	More	Not more	More
Forgotten to take pills	81.8	18.2	95.1	4.9	74.0	26.0
Taken more pills than prescribed	93.5	6.5	98.4	1.6	97.3	2.7
Skipped taking pills	92.2	7.8	100.0	0	87.7	12.3
Adherence to pills is better than before	67.5	32.5	85.2	14.8	71.2	28.8
Taken pills at a lower dose than prescribed	100	–	95.3	4.7	97.3	1.4
Taken pills at a different time	64.9	35.1	95.1	4.9	98.6	1.4
Forgotten to eat meals	92.0	8.0	96.7	3.3	93.2	6.8
Ate more often than before	49.3	50.7	52.5	47.5	93.2	6.8
Skipped meals	82.7	17.3	86.9	13.1	50.7	49.3
Ate more healthier than before	54.7	45.3	52.5	47.5	84.9	15.1
Ate at different times from before	44.0	56.0	50.8	49.2	90.4	9.6
Are more than before	53.3	46.7	49.2	50.8	27.4	72.6

CF, Cystic Fibrosis; LTX, lung-transplanted patients.

Table 4

Physiotherapy and nebulizing treatment since the start of the COVID-19 pandemic, classified as not more or more than before the pandemic.

	Patients with CF <i>n</i> = 66 (%)		Parents of children with CF <i>n</i> = 73 (%)	
	Not more	More	Not more	More
Physiotherapy is been forgotten	75.8	24.2	84.9	15.1
I/my child does more physiotherapy than prescribed	81.8	18.2	84.9	15.1
I/my child skip(s) physiotherapy	70.8	29.2	93.2	6.8
I/my child do(es) better physiotherapy than before	67.7	32.3	67.1	32.9
I/my child do(es) physiotherapy less often than prescribed	69.2	30.8	75.3	24.7
I/my child do(es) physiotherapy at a different time	35.4	64.6	69.4	30.6
Nebulising has been forgotten	77.1	22.9	79.5	20.5
I/my child nebulize(s) more often than prescribed	77.1	22.9	79.5	20.5
I/my child skip(s) nebulizing	84.3	15.7	89.0	11.0
I /my child nebulize(s) better than before	52.9	47.1	83.6	16.4
I/my child nebulize(s) less than prescribed	87.1	12.9	74.0	26
I/my child nebulize(s) at a different time	40.0	60.0	83.6	16.4

Data for the lung-transplantation patients have been excluded due to a small sample size [*n* = 6].

CF, Cystic Fibrosis.

3.3. Home CF and LTX treatment

Table 3 summarizes the self-reported changes in oral medication and nutrition since the start of the COVID-19 pandemic. Little change in oral medication was reported, although over 30% of the adults (CF) reported that they took their pills better than before or at a different time. Few LTX patients reported to forget, skip or change the timing of their medication. This indicates to no change in their routine of medication intake, which is reassuring, considering the nature of their medication (immunosuppressant medication). Half of the adult patients (CF and LTX) reported that they ate more often or more healthily, be it at different times than before; conversely, nearly 50% of the parents reported that meals were skipped more than before. Most of the respondents (46% of CF patients, 50% of LTX patients and 72% of parents) reported to eat more than before the start of the pandemic.

Table 4 summarizes the frequencies of patient visits to and by the home physiotherapist and changes in physiotherapy and nebulizing treatment. Only six LTX patients reported that they underwent physiotherapy; their data are not included.

Of the adult patients (CF), 86% reported undertaking their physiotherapy routine independently. Most of the children continued their treatment with the home physiotherapist, although 10% of the parents reported that their children, all teenagers, undertook their physiotherapy routine independently.

Around 60% of the adult patients (CF) reported undertaking their physiotherapy routine at a different time of the day than before the pandemic, and 30% did their physiotherapy less often. Around 32% of the adult patients (CF) reported positive changes in the quality of physiotherapy and 47% in the quality of nebulizing. In the responses from parents, 32% reported that their child under-

took his/her physiotherapy better than before the lockdown, and 30% reported that the physiotherapy was undertaken at a different time of day.

3.4. Health protecting behaviour and CF related worries

Table 5 summarizes the responses for the items describing health protecting behaviour and CF related worries. During the lockdown, all children were required to stay at home. Most of the adult patients (CF and LTX) were given sick leave so they could stay safely at home. Nearly all the respondents reported that someone else did their shopping or went to the pharmacy on their behalf, with the exception of some parents who went to the pharmacy themselves. Facemasks were worn when outside by 35% of the adults patients (CF), 47% of the LTX patients and 74% of the children. Over 40% of the adults (CF and LTX) were more worried when they coughed, or more worried about their CF, and 36% of the LTX patients reported stockpiling their medication.

More frequent temperature checking was reported by 32% of the adult patients (CF) and 58% of the LTX patients. Parents reported that 35% of the children were given more vitamins. Many of the respondents reported the increased use of hand sanitizers. Around 35% of the adult patients (CF and LTX) and 54% of the parents reported cancelling appointments more frequently. Over 40% of the adult patients (CF and LTX) reported exercising more since the lockdown.

3.5. COVID-19 status

None of the respondents had undergone testing for the virus (supplementary Table 2). The study excluded CF patients who were

Table 5

Health protecting behaviours and CF related worries since the start of the COVID-19 pandemic, classified as yes and no.

	Patients with CF n = 76 (%)		CF with LTX n = 61 (%)		Parents of children with CF n = 73 (%)	
	no	yes	no	yes	no	yes
Social distancing						
I/my child always stay(s) home	9.3	90.7	13.1	86.9		100.0
Someone else does the shopping	19.7	80.3	13.1	87.3	1.4	98.6
Someone else goes to the pharmacist	28.9	71.1	19.0	81.0	71.2	28.2
I/my child wear(s) a face masque when outside	64.5	35.5	52.4	47.6	26.0	74.0
CF related worries						
I am more worried when I/my child cough(s)	52.6	46.4	54.0	46.0	78.1	21.9
I stockpiled medication for myself/for my child	80.3	19.7	58.7	41.3	94.5	5.5
I worry more about my CF/my child's CF	46.1	53.9	63.5	36.5	79.5	20.5
Health protecting measures						
I check my/child's temperature more often	67.1	32.9	41.3	58.7	90.4	9.6
I/my child take(s) more vitamins	90.8	9.2	88.9	11.1	64.4	35.6
I/my child use(s) more hand sanitizer	19.7	80.3	15.9	84.1	63.0	37
I have cancelled my/my child's hospital appointments	64.5	35.5	73.0	27.0	45.2	54.8
I/ my child exercise(s) more	52.6	47.4	54.0	46	76.7	23.3

CF, Cystic Fibrosis; LTX, lung-transplanted patients.

hospitalized; however, it is known that all were tested before admission, and none tested positive.

3.6. Lung function, BMI, change in treatment and self-reported emotional impact

We hypothesized that lower FEV₁%pred, lower BMI (or BMI z-score) or CLAD status would be associated with a greater emotional impact. However, no associations were found for BMI, BMI z-scores or CLAD status, and, contrary to the hypothesis, the results showed that for the adult patients (CF), higher FEV₁%pred was associated with greater change with some of the items assessing emotional impact. Higher mean FEV₁%pred was associated with these patients reporting more stress, more negative thoughts about the crisis, more irritation about developments around COVID-19 and greater fear of contaminating others (supplementary Table 3).

Hypothesis 2, that lower FEV₁%pred and BMI (or BMI z-score) of CLAD status would be associated with greater change in treatment, was not confirmed for the adult patients (CF and LTX). However, the parents' responses showed a significant change in nebulizing therapy: in comparison to the group of parents of children with higher FEV₁%pred (mean, 100.8%; SD, 15.9%), the parents of 11 children with lower FEV₁%pred (mean, 85.5%; SD, 11.8%) reported that 'nebulising has been forgotten', but also improved nebulizing and nebulizing at a different time ($p < .01$).

4. Discussion

This study explored the early impact of COVID-19 on the emotional well-being of patients with CF, including those who had undergone LTX and paediatric patients, and recorded changes in home therapy since the start of the pandemic. The response rate (63%) was good, especially considering this was an online questionnaire, for which response rates are generally lower than for postal or telephone questionnaires [10]. It is important to note that with the start of COVID-19, the hospital started a campaign to promote the use of the Mynexuzhealth application, through which the questionnaire was sent.

The quick and good response rate may reflect the respondents' concerns and willingness to share their experiences and thoughts, their support for the initiative and a good relationship with our CF team. Some may have had more free time, but many were working from home and/or attempting to school their children.

None of the respondents had contracted COVID-19, indicative to the assumption that the study reports on a well characterized population, and is representative of many nation's low incidence of COVID-19 in CF at that time.

When considering the results, one has to keep the following quote in mind: "The traumatising character of an emotionally disturbing event is always the result of a personal and subjective interpretation of this event by the individual struck by the event and not merely dependant on objective cues in the given event" [11].

4.1. Emotional impact

The responses to the lockdown and COVID-19 pandemic were consistent with the findings reported by Brookes et al. in healthy people [3]. Changes in physical reactions were not reported by many of the respondents, apart from tiredness at the end of the day. The impact of the lockdown was reflected in respondents reporting more sadness, discouragement, feelings of helplessness regarding family or friends, greater criticism of others, perceptions of deterioration of the situation and difficulty with adhering to their routine. With regard to the items about the symptoms of posttraumatic stress, the respondents reported more stress, negative thoughts, and difficulty sleeping or concentrating; in addition, they felt more alert towards potential dangerous situations and were irritated by unexpected circumstances related to the crisis.

Patients with CF are a high-risk population, so it was not a surprise to find an increased fear of infection with COVID-19, or fear about infecting others. We can not conclude from our data that the psychological impact is different and specific in CF patients as compared to the general population. Fear of transmitting the virus, fear of contracting the virus and higher levels of alert may cause similar psychological reactions in all subjects experiencing this unprecedented pandemic. But the results are more worrisome in patients with CF, especially if this impact is sustained. The baseline incidence of anxiety and depression is elevated in these patients and parents [5] and preventive action and psychological treatment will have to take such findings into consideration.

The expected association between emotional impact and lung function was only partly confirmed and in the opposite direction to that hypothesised before the study. The adult patients (not LTX) with higher lung function reported greater stress, more negative thoughts about the crisis, more irritation toward developments related to COVID-19 and greater fear of contaminating others. It is possible that those facing worse severity of disease may have a generally increased baseline resilience over those who have milder disease. An other explanation may be that the patients with higher lung function experienced a greater impact because they have different responsibilities (work, financial etc.), than those with poorer lung function. Consequently, the lockdown prevented them from acting on these responsibilities; the future may therefore seem dis-

couraging. In addition, some patients with higher lung function may be more fearful, because they have put greater effort into their treatment regimen to achieve these results and want to sustain them. With this in mind, an external factor like COVID-19 is external to them and out of their control, potentially causing more fear. More research is needed to better understand the dynamics of these findings.

Conversely, the responses also showed signs of resilience. Only a few respondents reported loss of confidence, a loss of interest in things or degradation in taking decisions, which are all important characteristics for coping with a crisis. Many patients and parents commented on the lockdown and governmental regulations. Their stories indicated to their confidence in coping with the crisis at hand. For example: “We are used to wearing masks and washing hands and looking after our health” and most LTX patients commented: “Not so much has changed for me, I am always very careful and keep my distance”.

4.2. Home CF and LTX treatment

The majority of respondents reported that they forgot their treatment (unintentional non-adherence to treatment) more often since the start of the lockdown. This may be explained by the changes to their daily routine during self-isolation. The most often reported *intentional* changes in therapy were different timing of taking medicines (except amongst the LTX patients), timing of physiotherapy or nebulizing. Many parents specifically added a comment that they tried to keep to their usual routine as much as possible. Few changes in oral medication were reported, especially amongst the LTX patients, who are supposed to take their medication at set times.

Since the start of the lockdown more than half of the patients and parents reported eating extra, more often, more healthily, and at different times of day. They were encouraged by our dieticians, who sent out high caloric recipes on a weekly basis. These findings on changes in diet are promising, showing motivation for self-care, and it is to be hoped that these changes will be reflected in improvements in the weight and height (of the children) in the near future.

Plenty of time and perhaps fear of the virus may explain the finding that many adult patients and one third of the children changed the quality of their physiotherapy. Adults (CF) also reported a change in the quality of their nebulising therapy.

Overall, the changes in home treatment reported by more than 30% of the respondents were encouraging, indicating resilience in self-management in difficult circumstances. The challenge for both patients, parents and CF teams, is to maintain these positive changes over time and promote resilience when not faced with an international pandemic.

4.3. Health protecting behaviour and CF related worries

The respondents reported staying at home during the lockdown and increasing specific health protecting measures, such as the increased use of hand sanitizers. Some findings were different between patients and parents. For example, the adult patients (CF and LTX) reported more worries about their CF and about coughing, but the parents did not. This may have been because the children were at home, which should be the safest place for them. Also, adult patients had more advanced CF and may consider themselves more at risk compared to parents considering their children at risk.

The adult patients (CF and LTX) reported exercising more than before the start of the pandemic; this may have been because they had more time to do so, but also because they were motivated to keep fit during the lockdown. The physiotherapists of our CF centre

sent out weekly exercise programs, to encourage the patients to keep exercising.

Especially worrying was the finding that some of the adult patients (CF) and half of the parents reported cancelling their hospital appointments. Of course, at the start of the crisis, hospital appointments were cancelled by the CF team, but this was only for about 2 weeks. When asked why the patients and parents would not attend their appointment they replied: “The hospital is a dangerous place where COVID-19 patients are hospitalized”. Cancelling routine outpatient clinic visits is worrisome and needs to be addressed to prevent undiagnosed exacerbations or other CF related problems being overlooked.

Soon after the start of the lockdown videoconsultations were put in place, opening up the possibility of CF telehealth care. Weekly newsletters provided information on the hospital's preventative measures to assure safety for patients. We specifically asked for the patients' ideas about their CF care. An important question was: “When do you think you will dare to come to the hospital again?” This often triggered a positive discussion, as most patients and parents know they need to attend the CF clinic at some point.

4.4. Limitations

This study had several limitations, of which the most important was the ad hoc development of the questionnaire in the absence of a validated instrument. The items on impact were based on existing questionnaires, but it was not possible to test for reliability and validity. With the lack of psychometric properties, multivariate analyses were not considered valid. Similarly, there were no baseline values or comparison data from healthy individuals or patients with other chronic diseases, and we only have the respondents' view since the start of the pandemic. This self-report has its' limitations, including social desirability and over-estimation. In addition, we did not have the children's own ratings of the impact of the pandemic.

A limitation of online questionnaires is the reluctance to use them because of safety and confidentiality issues [10]. To prevent this, the online questionnaire in this study was delivered via a secure application that is linked to the hospital's electronic medical patient records. Still, other limitations of online questionnaires need to be considered when interpreting the results, including concerns regarding the reliability and validity of the data obtained [10].

The time during which the respondents completed the questionnaire corresponded with a peak of the pandemic in Belgium (April 2020). A follow-up study is needed to examine the incidence of lasting symptoms of post-traumatic stress within this population, further changes in home treatment, and how well these patients cope with the situation. Mental health research is definitely a research priority for the COVID-19 pandemic [12].

4.5. Conclusion

The current pandemic is likely to last for some time. This has implications for the care of patients with CF and LTX, including a need for the provision of psychosocial support to help reduce levels of stress and anxiety. The findings of this study on home CF treatment were promising; they suggest that focus should be given to the further motivation of patients and parents to improve their self-management. It is also important to contextualize the information that patients and parents receive from the media and to support them in balancing perceived risk and true risk. There is a crucial role for multidisciplinary teams, which can pro-actively connect with, inform and support the patients and parents.

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Declaration of Competing Interest

None.

CRediT authorship contribution statement

Trudy Havermans: Conceptualization, Methodology, Formal analysis, Investigation, Resources, Data curation, Writing - original draft, Writing - review & editing, Visualization, Supervision. **Janne Houben:** Conceptualization, Methodology, Software, Formal analysis, Investigation, Resources, Data curation, Writing - original draft, Writing - review & editing, Visualization. **Francois Vermeulen:** Conceptualization, Resources, Writing - original draft. **Mieke Boon:** Conceptualization, Resources, Writing - original draft. **Marijke Proesmans:** Conceptualization, Resources, Writing - original draft. **Natalie Lorent:** Conceptualization, Resources, Writing - original draft. **Erik de Soir:** Conceptualization, Methodology, Resources, Writing - original draft, Writing - review & editing. **Robin Vos:** Conceptualization, Resources, Writing - original draft. **Lieven Dupont:** Conceptualization, Methodology, Resources, Writing - original draft, Writing - review & editing, Supervision.

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Supplementary materials

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